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SPONTANEOUS RUPTURE OF RENAL ANGIOMYOLIPOMA

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We report a case of renal angiomyolipoma with retroperitoneal hemorrhage treated by enucleation in a 47 year-old male. The mass in the anterior side of the left kidney, revealed by sonography and CT, was diagnosed as angiomyolipoma with a retroperitoneal hematoma caused by its spontaneous rupture. Removal of hematoma and enucleation of the tumor were performed after the diagnosis.

Diagnosis and treatment of ruptured renal angiomyolipoma are discussed.

Key words: Retroperitoneal hemorrhage, Renal angiomyolipoma, Enucleation

INTRODUCTION

Renal angiomyolipoma (AML), which can be easily diagnosed by imaging, is dealt with mainly by conservative treatment[1,2]. However, when there is spontaneous rupture, the kidney is seldom preserved probably because of certain emergency features involved[3].

We report a case of renal AML accompanied by spontaneous rupture, a characteristic complication, and treated by enucleation. We have also reviewed the relevant literature.

CASE REPORT

The patient was a 47-year-old male truck driver. He was admitted to a neighboring hospital for severe left flank pain. He had previously passed a left ureter stone. Sonography indicated a mass on the anterior side of the left kidney. The following day he was transferred to our hospital and was suspected of having aneurysmal rupture.

There was no adenoma sebaceum on the face. The abdomen was swollen, and a mass the size of a child's head was detected along with acute tenderness in the left flank. Decreased intestinal peristalsis was noted. Peripheral blood examination showed leukocytosis (WBC 15,600/mm) and anemia (RBC 275 10/mm, Hb 8.6 g/dl, Ht 26.6%). A biochemical test indicated total protein of 5.4 g/dl, lactic dehydrogenase, 340 IU/l, creatinine, 1.4 mg/dl, blood urea nitrogen, 29.6 mg/dl. Urinalysis showed no abnormalities. Drip infusion pyelography (DIP) confirmed slight dilatation of left renal calices and pelvis and they were slightly compressed from below (Fig. 1, left). Sonography indicated on the ventral side of the lower pole of the left kidney, an echogenic tumor with a partialy transsonic area at the center as

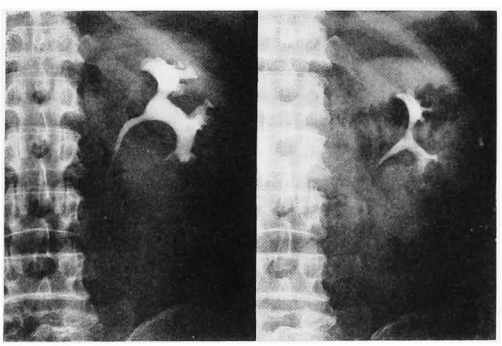

Fig. 1. Left: Preoperative DIP shows slight dilatation of left renal calices and pelvis, and their slight compression from below. Right: Postoperative DIP shows no dilatation of renal calices or pelvis.

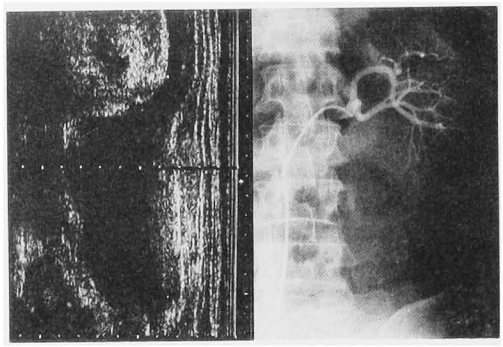

Fig. 2. Left: Preoperative sonography indicates, on the ventral side of the lower pole of the left kidney, an echogenic mass having a partialy transsonic area at its center as well as a transsonic area extending from the circumference of the tumor to the iliac crest. Right: Selective renal arteriography shows a tortuous coarse, feeding vessels at the area corresponding to the lesion of the lower pole, but there is no indicating bleeding.

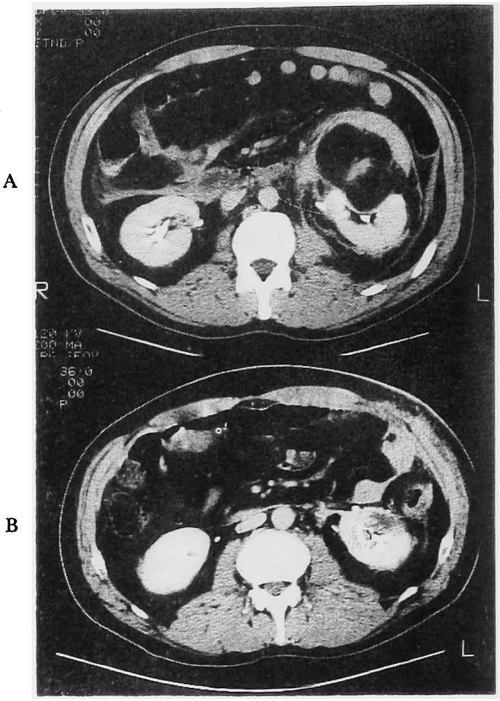

Fig. 3. A: Preoperative CT scan indicates, on the ventral side of the lower pole of the left kidney, a low absorption lesion of about 8 cm-diameter, having a high absorption area extending from the circumference of the lesion to the iliac crest. B: Postoperative CT scan shows a partial area of insufficient contrast on the lesion, and no residual tumor is evident.

well as a transsonic area extending from the circumference of the tumor to the iliac crest (Fig. 2, left). Computed tomography (CT) indicated, on the ventral side of the lower pole of the left kidney, a low absorption lesion of about 8 cm in diameter, with a high absorption area extending from the circumference of the lesion to the iliac crest (Fig. 3, A). These findings suggested left renal AML with retroperitoneal hemorrhage due to spontaneous rupture. Subsequent selective renal arteriography (AG) showed a tortuous coarse feeding vessel in the area corresponding to the above lesion of the lower pole, but no indication of bleeding. Thus embolization was not necessary (Fig. 2, right).

The anemia was improved by conducting a 5-unit blood transfusion, and the operation was performed ten days after its onset. Laparotomy by left subcostal arched incision indicated a hematoma the size of a child's head along the aorta in the retroperitoneum. After putting a clamp on the renal artery and vein, we exposed the anterior surface of the hematoma from the outside of the descending colon. It was removed and washed, in two steps. A yellow colored goose-egg size tumor subsequently became evident in the lower pole of the left kidney. A portion of the tumor was quickly diagnosed, and AML was confirmed pathologically. The tumor was enucleated. (Ischemic interval, 33 min; blood loss including hematoma, 1,904 ml; operating time, 195 min).

Histologically, the tumor tissue consisted of adipose tissue, rich in blood vessels, with sporadical smooth muscle cell hyperplasia perivascularly. Constitutive cells showed neither atypical nor nuclear divisional imaging (Fig. 4).

Temporarily postoperative ileus occurred, but was relieved conservatively. Postoperative DIP showed no dilatation and the left renal calices and pelvis were outlined well (Fig. 1, right). CT revealed a area with poor contrast on the ventral side of the lower pole of the left kidney, but no residual tumor (Fig. 3A).

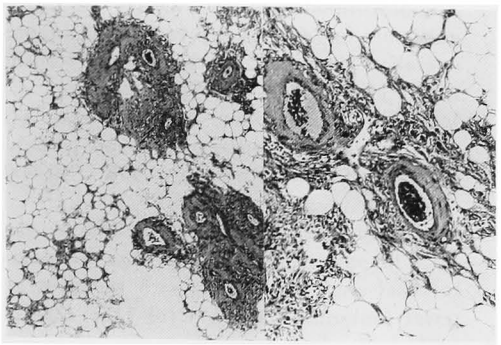

Fig. 4. Microscopic appearance of the tumor (left HE, ×44. right HE, ×130). The tumor tissue is composed of adipose tissue, rich in blood vessels, with sporadical smooth muscle cell hyperplasia perivascularly. Constitutive cells show neither atipical nor nuclear divisional imaging.

DISCUSSION

An increasing number of cases of renal AML have been reported owing to the improvement in imaging techniques such as CT and sonography. Now we find difficulty in regarding this disease as a very rare one. However, since we occasionally encounter cases which may rupture spontaneously or can be difficult to be differentiated from renal cell carcinoma, this disease still remains to be clinically a very important entity.

Spontaneous rupture of renal AML often occurs with flank pain in the affected side, nausea, vomiting, shock and other conditions and is included in the category of the so-called acute abdomen. Therefore, laparotomy is often performed under the preoperative diagnosis of perforated peritonitis due to appendicitis[4], torsion of ovarian cyst[5]. and others, resulting in nephrectomy.

The incidence of spontaneous rupture of renal AML was reported to be 16.5% (32 out of 194 cases) by Amano et al.[3] in Japan and 14.2% (15 out of 105 cases) by Mounded et al.[6] in Europe and America. Oestering et al.[1], who classified renal AML according to tumor size, reported an incidence of 90 tumors with hemorrhagic sign out of 178 cases (51%) and that of 10 out of 75 cases (13%) above and below 4 cm. On the other hand, McDougal et al.[7] reported 78 cases of renal rupture associated with perirenal hematoma. Forty five cases (57.7%) were tumorous, and 26 of them were malignant (including 15 cases of renal cell carcinoma). They found 9 cases of renal AML among the remaining 19 cases of benign tumor. Therefore, although spontaneous rupture of the tumor is a characteristic finding, renal AML should not be regarded as the main underlying disease, but rather as a malignant tumor that should be given serious consideration.

In the case of an emergency operation as mentioned here, renal conservation is hardly possible even if renal AML is strongly suspected[2,5]. This may be mainly because renal AML can not be depicted accurately by the actual image diagnosis, as shown by the existence of those cases which have been difficult to distinguish preoperatively from renal cell carcinoma[8,9] and because spontaneous rupture itself of a renal tumor is rather among the findings suggestive of the malignancy.

We believe that when spontaneous rupture of renal AML is strongly suspected by sonography, CT and when the general condition of the patient is stable, we should try to avoid the emergency operation which is possible through such treatments transfusion and others after determining the site of bleeding by arteriography (embolization[10], if necessary). After these measures have been taken, we should proceed to the surgery and upon pathological determination of renal AML, should try to conserve the kidney, if possible, by partial nephrectomy[4] or enucleation[11], as in the case of renal AML without rupture.

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和文抄録

後腹膜出血にて発症した腎血管筋脂肪腫

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後腹膜出血を伴う腎血管筋脂肪腫にたいして，核出術を行いえた1例を報告した．症例は47歳男性で，左側腹部痛を主訴に入院した．画像診断上，腎血管筋脂肪腫とその破裂による後腹膜血腫がもっとも疑われた．迅速診断の上，血腫除去と核出術が行われた．

破裂を伴う腎血管筋脂肪腫の診断・治療等につき考察した．

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